CASE REPORT

Intravitreal cysticercosis with full thickness macular hole: management outcome and intraoperative optical coherence tomography features

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SUMMARY

Ocular cysticercosis is a serious condition with a potential for complete vision loss if left untreated. Intravitreal cysticercosis is the most common ocular form of cysticercosis and is associated with retinal detachment, retinal traction, subretinal scarring and vitritis. To the best of our knowledge, there is no report of the occurrence of a live intravitreal cysticercosis with a full thickness macular hole (FTMH) in the literature. We here report a case of live intravitreal cysticercosis with a FTMH along with its management and intraoperative optical coherence tomography features of the live cysticercus.

BACKGROUND

Cysticercosis in humans is caused by tissue infestation of the cysticercus larva of the pork tape worm *Taenia solium*.¹ Ocular cysticercosis is a serious condition with a potential for complete vision loss if left untreated.^{2,3} Intravitreal cysticercosis is the most common ocular form of cysticercosis and is believed to be caused by intraocular migration of the larva from the bloodstream through the choroid, subretinal space and neurosensory retina into the vitreous cavity.^{3,4} To the best of our knowledge, there is no report of management of intravitreal cysticercosis with a full thickness macular hole (FTMH) in the literature. We here report a case of live intravitreal cysticercosis with a FTMH and its management. We also report the intraoperative optical coherence tomography (iOCT) features of live intravitreal cysticercosis.

CASE PRESENTATION

A 34-year-old man with no ocular or systemic premorbidities presented to our tertiary care clinic, with the chief complaint of diminution of vision in the right eye for the last 6 months associated with a scotoma in the superior visual field. On evaluation, his best corrected visual acuity (BCVA) was 3/60, <N36 in the right eye (OD) and 6/6, N6 in the left eye (OS). Examination of the right eye also showed 0.5+ cells in the anterior chamber and 1+ cells in the anterior vitreous. Rest of the ocular examination including ocular movements, lids, adnexa and anterior segment was within normal limits in both the eyes. A cystic mass was seen in the anterior vitreous on slit lamp examination in down gaze in

OD. Intraocular pressures were 12 and 14 mm Hg in OD and OS respectively.

Slit lamp biomicroscopy OD showed the presence of a FTMH with regular margins, grade-0 epiretinal membrane and xanthophyll pigments at the base. A translucent cyst with an internal dense white structure suggestive of cysticercus with scolex was seen inferotemporally in the vitreous cavity (figure 1). The cyst showed surface movements on illumination with indirect ophthalmoscope. Rest of the fundus was unremarkable. Left eye examination was essentially within normal limits.

INVESTIGATIONS

Spectral domain optical coherence tomography (SD-OCT) (Spectralis HRA; Heidelberg Engineering, Heidelberg, Germany) confirmed a FTMH with minimum diameter of 740 µm and a macular hole index of 0.17 (figure 2). The ellipsoid zone and external limiting membrane on the elevated retina were found to be continuous but fell short of reaching the margins of the hole. The retinal pigment epithelium (RPE) showed bumpiness, corresponding to the yellow pigments seen clinically. Short wavelength autofluorescence showed two areas of focal hypoautofluorescence suggestive of focal RPE loss (arrowheads in figure 3). There was a focal area of RPE thickening seen on SD-OCT (arrow in figure 2) which corresponded to one of the areas of hypoautofluorescence seen on short wavelength autofluorescence (lower arrowhead in figure 3). B-scan ultrasonography (USG) revealed a well-defined cystic lesion in the vitreous cavity with clear contents and a hyperechoic area which showed high amplitude spikes on A-scan suggestive of a scolex. Orbital USG revealed no cystic lesions in the orbit or extraocular muscles.

Stool examination was normal. Blood examination revealed eosinophilia. There was no evidence of subcutaneous nodules in the body. MRI of brain showed multiple ring-enhancing lesions suggestive of neurocysticercosis.

TREATMENT

Patient underwent 23G pars plana vitrectomy, posterior vitreous detachment (PVD) induction, in vivo cyst lysis and aspiration, and brilliant blue G-assisted internal limiting membrane (ILM) peeling with inverted ILM flap over macular hole with 25% SF6 gas injection. Intraoperatively it

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Figure 1 Widefield imaging (Optos 200TX; Optos, Dunfermline, UK) showing a large macular hole with an intravitreal translucent cyst and a normal retinal periphery.

was noted that there was a small rim of epiretinal membrane formation around the margins of the hole which was negatively stained with brilliant blue G dye. iOCT (OCT system attached to the Zeiss OPMI Lumera 700/Rescan 700 microscope; Zeiss Ltd, Jena, Germany) was used to study the features of the cyst after PVD induction when the cyst reached a preretinal location (figure 4). It was also used to confirm placement of the inverted ILM flap over the FTMH. Postoperatively the patient was given facedown positioning for 1 week.

OUTCOME AND FOLLOW-UP

iOCT showed two layers of hyperechoic cyst walls with the outer cyst wall being more hyperechoic than the inner wall. There were cobweb-like reflections between the walls possibly suggesting the duct systems of the cysticercus wall (figure 3).⁵ There was an invaginated hyperreflective structure at one pole of the cyst suggestive of a scolex. At 1 month follow-up, the BCVA had improved to 6/36, N18 OD. Macular hole was closed with the ILM flaps on the macular hole visible on SD-OCT (figure 5).

DISCUSSION

Human infection with *cysticercus cellulosae* is a common problem in the developing countries and occurs due to ingestion of eggs of *T solium* or by reflux of gravid proglottids from lower intestines into stomach and their subsequent excystment.⁶ The embryos hatched out of the eggs penetrate the mucosa and via the portal venous system enter systemic circulation and lodge

Figure 3 Preoperative short wavelength autofluorescence (Spectralis HRA; Heidelberg Engineering) showing two focal areas (arrowhead) of reduced autofluorescence at the base of the hole indicating patchy loss of retinal pigment epithelial cells. The lower patch corresponds to the retinal pigment epithelium thickening noted in the spectral domain optical coherence tomography (arrow, figure 2).

at sites with high blood circulation such as the brain, eye, skin and skeletal muscles.^{7 8} In the ocular tissues, macula has been noted to be the preferred site for the lodgement of the blood-borne cysticercus possibly due to the rich blood supply.⁷ From the macular subretinal space it can enter the vitreous cavity through a break in the overlying neurosensory retina. When this migration occurs, the defect in the retina thus formed can give rise to rhegmatogenous retinal detachment, or more commonly this site heals with a scar due to the inflammation associated with the cysticercus migration and leads to an area of scarring in the retina.⁹ Sharma *et al* have hypothesised an alternate route of entry for cysticercus into the vitreous cavity based on their observation in four cases of intravitreal cysticercosis and no retinal lesion—through a retinal blood vessel, optic disc or ciliary body.³

Authors have reported a case of live intravitreal cyst communicating with the subretinal space demonstrating the larva entering the vitreous cavity through a break in the retina.¹⁰ George *et al* have also reported an intravitreal cysticercosis with an associated retinal break.¹¹ While macular area has been noted to be the most common region for lodgement of cysticercus

Figure 2 Preoperative spectral domain optical coherence tomography (Spectralis HRA; Heidelberg Engineering) showing a large macular hole with a basal diameter of 2309 µm. Arrow indicates a focal area of retinal pigment epithelium thickening.

Figure 4 Intraoperative optical coherence tomography (Rescan 700; Carl Zeiss Meditec, Germany) appearance of the cyst showing an invaginated scolex.

(about 80%),⁷ it is only rarely documented to exit via macula to reach the vitreous cavity. Kruger-Leite *et al* have reported a case of large macular break associated with escape of a submacular cysticercosis into vitreous cavity.⁹ This subsequently led to scarring of the macula and poor visual outcome. Kaliaperumal *et al* have noted a case of macular hole associated with an intravitreal cysticercosis in their case series but further information on the clinical features and subsequent management of the case is not presented.¹² To the best of our knowledge, there is no report of a patient presenting with a FTMH and an associated live intravitreal cysticercosis.

In our case, the cyst was present in the inferior and anterior vitreous, and there were no other abnormalities in the fundus examination to suggest any other site of entry of cysticercus into the vitreous cavity other than the macular hole. Also, the macular hole did not show any signs of traction on the hole or cystoid changes at the edges to suggest an alternative cause for macular hole formation. In addition, autofluorescence imaging showed areas of hypoautofluorescence suggesting focal RPE loss. We, therefore, hypothesise that the macular hole in this case was caused by the migration of the cysticercus into the vitreous cavity from a subretinal location. Our case also characteristically lacked notable active vitreous inflammation, retinal detachment, retinal traction, subretinal fibrosis or any other ominous features generally associated with intravitreal cysticercosis.

Figure 5 Postoperative spectral domain optical coherence tomography (Spectralis HRA; Heidelberg Engineering) showing a closed macular hole with inverted internal limiting membrane flaps visible on the hole.

OCT features of cysticercosis in the subretinal space, preretinal location and anterior chamber are reported.^{13–15} In this case, preoperative OCT was not possible due to the location of the cyst which could neither be imaged using an anterior segment OCT or a posterior segment OCT. iOCT helped imaging the cyst intraoperatively. iOCT demonstrated the internal structure of the cysticercus better due to the possibility of rotating the cyst intraoperatively and taking sections through any desired region of the cyst. Examination of the iOCT images showed close correlation with the described histopathology of the cysticercus.⁵ iOCT also helped us to confirm the placement of inverted ILM flaps over the macular hole and confirm its presence on the macular hole after fluid–air exchange.

Learning points

- ▶ Intravitreal cysticercosis can present with an associated full thickness macular hole.
- ▶ Examination of the blue autofluorescence image from macula suggests that the macula may be the site of exit of cysticercus from the subretinal space.
- ▶ Timely intervention in these cases can preserve visual function.

Contributor KR: data acquisition, analysis and interpretation of data, manuscript preparation. RDR: data acquisition, manuscript preparation, critical revision of the manuscript. PK: manuscript preparation and critical revision of the manuscript. AK: conception and design of the work, critical revision of the manuscript.

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